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Primary thyroglossal duct cyst surgery: A UK tertiary referral paediatric centre experience

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Abstract

Objectives

To report the clinical outcomes following primary thyroglossal duct cyst (TGDC) surgery.

Methods

Retrospective case note review of 79 patients undergoing surgery for suspected TGDC at our tertiary referral paediatric centre, excised between July 2014-November 2018.

Results

TGDC was confirmed histologically in 57.0%. Classic Sistrunk procedure (CSP) was performed in 60.0%, modified Sistrunk procedure with central neck dissection (MSPCND) in 35.6% and cystectomy in 4.4%. Overall recurrence was 15.6%. Of those who recurred, 85.7% had CSP and 14.3% had cystectomy. There was no recurrence in those who had MSPCND. Post-operative infection occurred in 14.8% undergoing CSP, 12.5% undergoing MSPCND and 0% undergoing cystectomy. Return to theatre was required in 7.4% undergoing CSP and 0% who had MSPCND or cystectomy. None experienced post-operative haemorrhage.

Conclusion

MSPCND for primary TGDC was not associated with any increase in morbidity compared with CSP and appears to have a lower risk of recurrence.

Keywords

Thyroglossal cyst, otolaryngology, radiology

Introduction

Thyroglossal duct cysts (TGDC) are the most common cause of midline neck lumps in children.⁽¹⁾ They account for 70% of all paediatric neck swellings⁽²⁾ and occur in approximately 7% of the population.⁽³⁾ During gestation, the thyroid gland descends from the foramen caecum at the junction of the anterior 2/3 and posterior 1/3 of the tongue and passes very close to the hyoid bone before settling on the anterior trachea.⁽⁴⁾ The fibrous tract connecting the thyroid to the foramen caecum obliterates under normal circumstances. However, it occasionally persists as a thyroglossal duct allowing the formation of mucus filled cysts within it.⁽⁵⁾ As such, the cyst presents as a midline neck mass that moves on deglutition and protrusion of the tongue, which often yields the clinical diagnosis.⁽⁶⁾ Very rarely the cyst may represent an ectopic thyroid or contain the only thyroid tissue in the body⁽⁷⁾ and surgical excision would inadvertently cause hypothyroidism necessitating lifelong medication. Therefore, an ultrasound scan is usually requested to differentiate these pathologies.⁽⁸⁾

Surgery is recommended for preventing recurrent infection and cervical fistulisation⁽⁹⁾ as well as providing a histological diagnosis, as there is a 1% risk of carcinoma.⁽¹⁰⁾ In 1920, Sistrunk⁽¹¹⁾ presented his procedure based on the embryology of the disease. It is eponymously known as the Sistrunk procedure (SP), which involves excising the cyst, the body of the hyoid and a core of tissue up to and including the foramen caecum without breaching the oral mucosa. Using this method the recurrence rate had reduced from 40% to 5%. Histological studies have shown that the main duct may have many microscopic lateral, superficial and deep branches, which may give rise to recurrent cysts if inadequately excised.⁽¹²⁾ Based on these findings, several authors have described a modification of the SP with the addition of a wide central neck dissection (CND) to excise the tract and all possible branches *en bloc*.^(13,14)

The main challenges involved in this condition include 1) diagnosis as typical clinical signs may be absent and radiology may be inconclusive and 2) recurrence of disease in those who have undergone surgical intervention.⁽¹⁵⁾ We report the clinical outcomes following surgery for TGDCs performed at our tertiary referral paediatric centre.

Materials and methods

Our local audit department provided a list of patients who had undergone surgery for suspected TGDC between July 2014 and November 2018. Patients undergoing surgery before July 2014 were excluded due to difficulty in accessing case notes. This returned 79 patients, whose electronic notes were then reviewed for histological confirmation of TGDC. This revealed 47 cases of histologically confirmed TGDC and 32 cases in which the histology was either inconclusive or inconsistent with a diagnosis of TGDC.

This created two study groups – A TGDC group and non-TGDC group. In the TGDC group, 2 were recurrent TGDCs and were excluded form the study. The total final number was 45 in the TGDC group and 32 in the non-TGDC group (Figure 1). Whilst 2 groups were created, the main focus of this study was the outcomes relating to those confirmed to have TGDC. However, important results identified from the non-TGDC group have also been presented.

Data was extracted from electronic case notes including demographics, symptoms, pre-operative investigation results, surgery performed, post-operative complications, post-operative antibiotic administration, histology, recurrence and final outcome. The sensitivity, specificity, positive predictive value (PPV) and negative predictive value (NPV) of radiological evaluation were calculated.

Procedures performed in our series included classic Sistrunk procedure (CSP), modified Sistrunk procedure with central neck dissection (MSPCND) and cystectomy. Definitions of these procedures documented in the literature are inconsistent. In our series, we defined 1) CSP as resection of the cyst, body of the hyoid with or without a core of suprahyoid soft tissue and a wedge of tongue base musculature, the amount of which can vary up to the foramen caecum without breaching the oropharyngeal mucosa, 2) MSPCND as resection of the same structures as in CSP but including a cuff of strap musculature and resulting *en bloc* resection of the cyst, straps, hyoid and tongue base wedge and 3) cystectomy as cyst excision alone.

Results and analysis

Demographics

The mean patient age was 6. There were 28 males and 17 females with a male:female ratio of 1.6:1. The mean follow up period was 6.6 months with a range of 2 weeks – 58 months.

Symptomatology

All cases presented with a midline neck swelling. 9/44 (20.5%) presented with pain or tenderness, 7/44 (15.9%) with sinus or discharge and 21/43 (48.8%) with preceding history of infection. None of our patients had dysphagia or airway symptoms. Data for 1/45 (2.2%) was unavailable. The mean duration of symptoms before presentation was 8.1 months. Movement on swallowing was observed in 9/14 (64.3%) (data unavailable in 31/45) and on tongue protrusion in 14/19 (73.7%) (data unavailable in 26/45).

Diagnostic work-up

Pre-operative investigation results were unavailable in 2/45 (4.4%). Pre-operative ultrasonography was performed in 42/43 (97.7%), thyroid function tests in 6/43 (14.0%), magnetic resonance imaging (MRI) in 4/43 (9.3%), nuclear medicine imaging in 1/43 (2.3%) and fine needle aspiration in 0/43 (0%).

Radiological Evaluation – sensitivity, specificity, PPV and NPV

In the TGDC group, radiological examination confirmed TGDC in 32/43 (74.4%) scans. The findings of the remaining 11/43 (25.6%) cases are demonstrated in table 1. In the non-TGDC group there were 32 cases. Within this group, radiology results were unavailable for 1/32 and were excluded. Radiology suggested TGDC as the most likely diagnosis in 17/31 (54.8%) and other diagnoses as more likely in 14/31 (45.2%).

Therefore, the sensitivity and specificity of the radiological evaluation of TGDC was 74.4% and 45.2% respectively. The positive predictive value (PPV) and negative predictive value (NPV) were 65.3% and 56.0% respectively (table 2).

Procedures performed and intra-operative findings

TGDC surgery was performed by ENT surgeons in 35/45 (77.8%), paediatric surgeons in 9/45 (20.0%) and plastic surgeons in 1/45 (2.2%). CSP was performed in 27/45 (60.0%), MSPCND in 16/45 (35.6%) and cystectomy in 2/45 (4.4%). Cysts were located in the midline in 22/45 (48.9%), right paramedian in 15/45 (33.3%) and left paramedian position in 8/45 (17.8%). Cysts were suprahyoid in 20/45 (44.4%) and infrahyoid in 25/45 (55.6%). The hyoid bone and wedge of tongue base musculature were excised in 42/44 (95.5%) (data unavailable in 1/45) and 26/42 (61.9%) (data unavailable in 3/45) cases respectively. The cyst was ruptured at surgery in 6/42 (13.0%) (data unavailable in 3/45).

Post-operative complications

Post-operative complications included 6/45 (13.3%) with infection and 2/45 (4.4%) required a return to theatre. The indication for return to theatre in both cases was post-operative infection. There were no cases of post-operative haemorrhage. Table 3 demonstrates the rate of specific post-operative complications according to type of procedure performed.

Post-operative antibiotic prescription

Data was unavailable for 2/45 (4.4%). Post-operative antibiotics were prescribed in 24/43 (55.8%). Of those prescribed antibiotics, 1/24 (4.2%) developed post-operative infection compared to 5/19 (26.3%) of those who did not. Of the 2/45 (4.4%) who returned to theatre for infection, neither received post-operative antibiotics. Figure 2 demonstrates the rate of post-operative infection between different procedures for those who did and did not receive antibiotics. Those who received post-operative antibiotics antibiotics antibiotics. The post-operative antibiotical therapy received between 5 to 7 days of oral co-amoxiclav.

Disease recurrence

Recurrence occurred in 7/45 (15.6%). Of the 7 who recurred, 6/7 (85.7%) had undergone CSP and 1/7 (14.3%) cystectomy. Of the CSP cases that recurred, the hyoid was excised in 6/6 (100%) of cases whilst a wedge of tongue base musculature was excised in 3/6 (50%) of cases. The only other patient undergoing cystectomy in the TGDC group did not recur. In all 16 patients who underwent MSPCND, none experienced recurrence. A revision operation was required in 6/7 (85.7%). A 2^{nd}

recurrence occurred in 2/6 (33.3%), one of which was the cystectomy case that, for the 2^{nd} time, underwent a procedure sparing the hyoid and tongue base musculature and ultimately underwent a MSPCND as a 3^{rd} procedure. They are still under review.

Outcome

Of the total number of patients, thirty-seven (82.2%) were discharged, 5/45 (11.1%) are still under review and 3/45 (6.7%) were lost to follow up.

Non-TGDC group – histology, radiology and procedures

In the non-TGDC group, 5/32 histology reports were unavailable. Of the reports available, 25/27 radiology reports were inconsistent with the final histology. Table 4 demonstrates the radiological diagnoses in this group.

15/32 (46.9%) in this group had a CSP. Of these, data was unavailable for 2/32 (6.3%). Therefore, excluding these, 15/30 (50.0%) had a CSP. Radiology reports were available for 9/15 (60.0%) and of these, 5/9 (55.6%) suggested TGDC as the most likely diagnosis.

8/32 (25.0%) in this group had a MSPCND. Data was unavailable for 2/32 (6.3%). Therefore, excluding these, 8/30 (26.7%) had a MSPCND. Radiology reports were available for 6/8 (75.0%) and of these, 4/6 (66.7%) suggested TGDC as the most likely diagnosis.

Discussion

Synopsis of key/new findings

There were no cases of recurrent TGDC following MSPCND. This procedure is associated with reduced rates of post-operative infection and return to theatre compared to CSP. Radiological evaluation cannot accurately diagnose TGDCs. Post-operative infection was lower in those who received antibiotics compared to those who did not.

Study Strengths

This study included patients presenting to a tertiary referral centre over a 4-5 year period and undergoing a procedure of interest to surgeons from different specialties. Due to the diversity of surgical practice in our hospital, we were able to compare outcomes between different surgeons, procedures and post-operative management.

Study Limitations

This study was retrospective and therefore no causal analysis could be performed. Data in some instances was incomplete due to unavailability of some patient health records. No statistical analysis was performed so we are unable to comment on the statistical significance of the results.

Average length of follow up was 6.6 months, which may limit our knowledge of recurrence or other complication in our population. Similarly, data was collected up until the date of the most recent clinic data. More recent cases will have a reduced follow up period compared to older cases, which may introduce lead-time bias.

Our data included different specialty surgeons performing a number of different procedures. Experience of the operating surgeon and a lack of standardisation in the procedure performed may introduce bias. Despite defining our procedures in this paper it was clear from case note review that there was variation within how CSP and MSPCND were performed from surgeon to surgeon. This was corroborated by a review of the literature, which revealed varying descriptions of CND. It is therefore inappropriate to draw firm conclusions from this study by comparing our results to those in the literature. However, we have commented on this as studies on this subject are limited.

Whilst our study includes one of the largest published series of MSPCND for primary paediatric TGDCs, it still has a limited sample size and will require further prospective study with a larger patient cohort.

Comparisons with other studies

Clinical outcomes following surgery for paediatric TGDCs are well documented.^(16,17) This literature review will therefore focus on studies relating to outcomes following CND.

Ahmed et al.⁽¹⁸⁾ reported no recurrence in 23 primary paediatric TGDCs undergoing an MSPCND. They performed the procedure in a further 15 recurrent paediatric cases and reported only one recurrence in this subgroup.

Two studies have reported data on CND for primary simple (non-infected) TGDCs. In 1986, Howard et al⁽¹⁴⁾ reported no increase in morbidity following CND for 14 TGDCs. No data regarding demographics or recurrence were given. More recently, Ubayasiri et al⁽¹⁹⁾ demonstrated no recurrence following CND in 12 patients. Demographic data was not provided.

A further two studies by Kim et $al^{(13)}$ and Isaacson et $al^{(20)}$ reported no recurrence following CND for children with primary complex (infected) or recurrent TGDCs.

Five studies have published data regarding CND for recurrent TGDC only. Pucher et $al^{(21)}$ investigated 73 patients who had either a CND or a modified Sistrunk procedure (MSP). They demonstrated recurrence in 1/45 and post-operative haematoma in 2/45 who had a MSP and no post-operative complications following CND. Whilst Pucher states CND is a safe procedure, they recommended its use for revision cases only due to risk of carotid artery, vagus nerve and laryngeal injury. These complications were not observed in our study. Similarly to Pucher et al, there were no recurrences reported by O'Neil et al⁽²²⁾ in 7 adults, Yao et al⁽²³⁾ in 28 children, Pastore et al⁽²⁴⁾ in 7 children and Mickel et al⁽²⁵⁾ in 20 patients.

In comparison, two authors had less success with CND in recurrent paediatric TGDCs. Perkins et al⁽²⁶⁾ reported recurrence in 33.3% and Ibrahim et al⁽¹⁵⁾ in 20% of children. However, of interest, they both found 0% recurrence following a suture guided transhyoid pharyngotomy procedure.

The vast majority of the literature corroborates our recurrence rate following CND. However, most studies report on complex primary or secondary TGDCs with only a few reporting outcomes for simple primary disease. Few studies evaluated other postoperative complications such as haemorrhage or infection. Only six authors overall investigated children specifically. All studies included a limited sample size.

Clinical applicability of the study

This paper includes the second largest published series investigating the effectiveness of MSPCND for primary paediatric TGDCs. Data regarding this area of research is limited and it is our hope that this study encourages further work in this area.

MSPCND was associated with a low complication rate and did not result in any disease recurrence compared to CSP. This should be taken into consideration when managing TGDCs as it may help to prevent disease recurrence without compromising post-operative morbidity.

Radiological investigations are requested to confirm the presence of a normal thyroid gland when a clinical diagnosis of TGDC has been made. However, in our series, these investigations suggested alternative diagnoses in many cases, which may have influenced surgical decision-making. This encouraged us to evaluate the accuracy of diagnostic radiology in the pre-operative workup for TGDC. We found that radiological evaluation for TGDCs had a sensitivity of 74.4%, specificity of 45.2%, PPV of 65.3% and NPV of 56.0%. In the non-TGDC group, 23/32 (71.9%) had either a CSP or a MSPCND. In these cases, radiology results were positive for TGDC in 17/31 (54.8%), which may explain why so many in this group had a Sistrunk related procedure. In the remaining 14/31 (45.2%) patients who had imaging suggestive of an alternative diagnosis, explanations for undergoing Sistrunk related procedures may include a strong clinical suspicion overriding imaging results or intentional aggressive surgery to prevent recurrent disease in the presence of diagnostic uncertainty. These findings suggest that imaging for paediatric TGDCs cannot be used in isolation with confidence when diagnosing TGDCs pre-operatively. Therefore, a full clinical evaluation should be used in conjunction with the imaging during surgical decisionmaking. Further studies are required to improve accurate pre-operative diagnosis of TGDCs.

Post-operative infection was lower in those who received antibiotics compared to those who did not. The two patients requiring a return to theatre for an infective complication did not receive antibiotics. These findings may suggest a benefit in routine antibiotic administration post-TGDC surgery. However, the potential benefit must be weighed against the deemed cost-effectiveness and risk of antimicrobial resistance.

Further study

Studies to improve the accuracy of pre-operative diagnosis of TGDCs are required. In addition, multicentre randomised controlled studies to assess the efficacy and safety of MSPCND in primary paediatric TGDCs and the role of post-operative prophylactic antibiotics would be helpful.

Conclusion

MSPCND for primary TGDC was not associated with any increase in morbidity compared with CSP and appears to have a lower risk of recurrence. Therefore, we suggest that this is the most appropriate surgical procedure for paediatric primary TGDCs. Peri-operative antibiotics appear to decrease the risk of post-operative infection. Radiological evaluation is not accurate in the pre-operative diagnosis of TGDC.

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Conflict of interest declaration

There is no conflict of interest to declare

Funding

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Ethical considerations

This study was registered with and approved by our local hospital audit department. We did not require ethics committee clearance as no new intervention was being studied.

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Bullet Point Summary

- To our knowledge, this study includes the second largest series of primary paediatric TGDCs undergoing modified Sistrunk procedure with central neck dissection (MSPCND)
- MSPCND may be more effective than CSP for preventing recurrent TGDC
- MSPCND was associated with a low complication rate
- Prophylactic antibiotics for TGDC surgery may reduce rates of post-operative infection
- Imaging is important in determining the presence of a normal thyroid gland pre-operatively. However, radiological diagnosis of TGDC was found to be unreliable in this study. Clinicians should consider the full clinical assessment in conjunction with the imaging results to minimise misdiagnosis, number of incorrect procedures performed and recurrence.

Diagnosis	Number
Suspected dermoid cyst	3
Comments on presence of thyroid gland only	2
Suspected branchial cleft cyst	2
No specific diagnosis reported	2
Suspected atypical infection	1
Suspected dermoid vs inflamed lymph node vs ectopic thyroid	1
Total number	11

Table 1. Alternative radiological diagnoses in the TGDC group

	TGDC positive (n=43)*	TGDC negative (n=31)**	Predictive Value
Radiology positive	32 (74.4%) ¹ True Positives	17 (54.8%) False Positives	32/49 (65.3%) ³
Radiology negative	11 (25.6%) False Negatives	14 (45.2%) ² True Negatives	14/25 (56.0%) ⁴

Table 2. 1) Sensitivity 2) specificity 3) PPV and 4) NPV of radiological evaluation in TGDC

Procedure	Infection	Haemorrhage	Return to theatre
CSP	14.8%	0%	7.4%
MSPCND	12.5%	0%	0%
Cystectomy	0%	0%	0%

Table 3. Rate of post-operative complications according to procedure performed

Pathology	Number
Dermoid	15
Histology unavailable	5
Inconclusive histology report	2
Recurrent TGDC	2
Inflammatory mass	1
Multiple dermoids	1
Inclusion epidermoid cyst	1
Non-specific ruptured cyst	1
Preformed cystic lesion without epithelial lining	1
Inflammatory cyst	1
Dermoid and thyroglossal cyst (dual pathology)	1
Branchial cyst	1
Inflammation and scarring	1
Ectopic cervical salivary gland cyst	1
Total Number	34

Table 4. Histopathology of the non-TGDC group



Figure 1. Formation of TGDC and non-TGDC groups



Figure 2. Rate of post-operative infection according to procedure undertaken with and without prophylactic antibiotics

Bullet Point Summary

- To our knowledge, this study includes the second largest series of primary paediatric TGDCs undergoing modified Sistrunk procedure with central neck dissection (MSPCND)
- MSPCND may be more effective than CSP for preventing recurrent TGDC
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- Imaging is important in determining the presence of a normal thyroid gland pre-operatively. However, radiological diagnosis of TGDC was found to be unreliable in this study. Clinicians should consider the full clinical assessment in conjunction with the imaging results to minimise misdiagnosis, number of incorrect procedures performed and recurrence.